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Phonological Alexia during Left Inferior Temporal Cortical Stimulation

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Abstract

Current cognitive models propose that reading can be achieved through phonological or lexical/semantic pathways. However, the anatomical networks subserving these processes remain largely unravelled. The objective of this report is to describe a case of isolated phonological alexia induced by electrical stimulation of the basal temporal cortex. A 19 year-old woman with refractory left temporal epilepsy was submitted to EEG monitoring with subdural electrodes and cortical stimulation as part of the evaluation for epilepsy surgery. The subdural electrodes were located in the basal temporal lobe, around the fusiform gyrus. Electrical stimulation of one of the electrodes induced reproducible alexia mostly involving pseudo words with visual, morphological and normalization errors. This pattern is consistent with phonological alexia. No other language, memory or visuo-spatial deficits were elicited. The epileptogenic area was surgically excised, sparing this functionally important region. The patient remains seizure free and no additional neuropsychological deficit was found after surgery. This represents the first case of isolated phonological alexia induced by a circumscribed lesion in the left inferior occipito-temporal cortex. Functional imaging studies suggested that this region of the brain harbours the visual word form area, an area selectively involved in reading. This case strengthens this possibility. Furthermore, it seems to indicate that the phonological pathway of reading segregates from the whole word lexico-semantic pathway already at this early stages of processing, in the basal occipito-temporal cortex. This case also suggests that neuropsychological testing of the basal occipito-temporal cortex in pre-surgical epilepsy patients should include tests for phonological alexia.

Keywords: Phonological alexia; Inferior temporal lobe mapping; Transient phonological alexia; Inferior temporal lobe mapping; Cortical stimulation

Introduction

Current cognitive models propose that two independent pathways can process reading, a lexical/semantic route that matches word form to meaning and a phonological/sublexical route whereby written symbols are converted to sounds [1]. Phonological alexia is a form of acquired alexia characterized by a selective difficulty in reading pseudowords and function words (i.e., words with no semantic representation), due to a phonological route impairment. No specific anatomical correlates have been associated to this type of alexia, for it has been observed in large left anterior peri-sylvian lesions in association with other cognitive deficits [2,3].

We report a case of transient and isolated phonological alexia due to electrical stimulation of the basal left occipito-temporal (LOT) cortex during the presurgical evaluation of an epilepsy patient. This case brings new insights into to function of this area and contributes to better presurgical planning of epileptic patients.

Case Report

A 19 year-old right handed girl, with 11 years of formal education, was admitted for epilepsy surgery planning. When 2 year-old, a left temporal lobe tumor that caused seizures was excised. At 14 year-old seizures recurred, characterized

by a sudden impairment of cognition, meaningless speech, bimanual and oroalimentary automatisms. Seizures became refractory to best medical treatment. Neurological examination disclosed a right homonymous hemianopia. Brain MRI showed a left anterior temporal lobe porencephalic cavity with T2 and flair hyper intensities in the most anterior, inferior and medial remnants of the temporal lobe, attributed to gliosis. The most anterior regions of the first, second and third temporal gyrus had been removed, but most of the fusiform and lingual gyrus were present, as well as part of the parahippocampal and hippocampal regions (Figure 1). The neuropsychological examination showed a mild impairment in verbal comprehension, visual naming of line drawings, and moderate deficits of attention and verbal and visual memory. On video-scalp EEG ictal onset occurred in posterior temporal leads. Subdural electrodes were implanted. One grid with 6 electrodes and another with 20 electrodes (4x5) were implanted over the basal LOT cortex, surrounding the fusiform gyrus, as confirmed by skull radiograph and direct visual inspection during surgery.

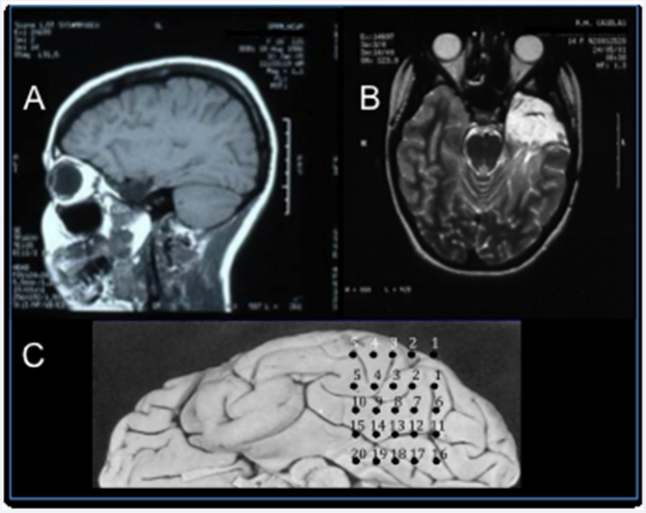

Figure 1: MRI and schematic representation of the electrodes location.

Electrical stimulation was applied to this region bipolarly. The electric current was increased from 0.5mA with 1mA steps, until after-discharges, clinical, electrical seizures or functional deficits were induced (stimulation threshold) or until a maximum intensity of 10mA. During stimulation the patient was asked to perform different tasks to assess language (picture description, auditory comprehension and naming of line drawings, colours and familiar faces), short-term memory and reading (reading aloud, reading comprehension though the classification of meaningful versus nonsense sentences). Whenever a reading difficulty was detected, further tests for reading pseudowords, words and numbers were used. Incorrect, delayed and non responses were rated as errors. All sessions were video recorded and electrocorticogram was recorded simultaneously. All tasks were also performed off-stimulation. Results are shown in (Table 1). There was a recurrent, consistent and selective reading deficit exclusively upon stimulation of electrode 6, located in the posterior aspect of the basal LOT, near the fusiform gyrus. The errors consisted mainly of paralexias: visual paralexias in phrases and word reading and regularization errors when reading pseudo words. There were also 4 hesitations in the beginning of the sentences and the patient could not read at all one sentence. 2 of the errors were non classifiable. Semantic paraphasias were not produced. Reading deficits were noted only in 3 trials out of 123 for the other electrodes. In one of these trials simultaneous repetitive epileptiform activity at electrode 6 was noted. The other two errors were pseudowords read with only a slight hesitation.

The patient was subsequently submitted to a surgical excision of the irritative and ictal onset zones, sparing the area surrounding electrode 6. The surgical procedure was unremarkable. After surgery, no reading disturbance was noted. At 2 years follow-up the patient is in Engel class Ia.

Discussion

To our knowledge this is the first report of an isolated alexia of the phonological type in western language caused by a circumscribed dysfunction of the fusiform gyrus in LOT region. Patients' performance is typical of phonological alexia because there was a predominant impairment in pseudo word reading (0/3) compared to words (8/13) and with lexicalization errors, transforming pseudo words into visually similar words. Other words were read with visual and morphological errors, all classically found in phonological dyslexia [2]. The particular clinical context limited the number of tests applied but the selectivity for pseudowords reading deficit and the inability to read small words (functors such as the word "a"), while sparing other complex visual stimuli and complex reading stimulus (such as complete sentences like "The telephone is used to talk"), can only be explained through a selective reading impairment.

The relevance of this report is threefold. It strengthens the selectivity hypothesis of the basal LOT area for reading. More importantly, it provides direct evidence for phonological processing in this area. For epilepsy care, it suggests the need to include phonological reading tasks in LOT pre-surgical mapping.

These claims are supported by functional neuroimaging literature and from lesional cases of Japanese readers. The LOT cortex, namely the left midfusi form gyrus (BA37), is consistently activated in fMRI studies of reading [4] and some authors locate the Visual Word Form Area (VWFA) within this region. That area responds selectively to words and pseudo words enabling a rapid perception of written words. However, there is still considerable debate regarding the selective role of these areas in reading.

Phonological dyslexia in western languages was described in associations with other deficits in patients with poorly localizing posterior lesions; pseudo words reading deficits were associated with strokes involving the middle frontal gyrus, the inferior

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frontal gyrus, the angular gyrus and the middle occipital gyrus [5]. In western language, it has not been shown that different reading pathways can be differentiated in the LOT region. Japanese has a dual system of writing/reading-Kana, grossly equivalent to western characters; and Kanji, where characters are invariably associated with meaning. In Japanese readers some reports have shown that lesions in the fusiform gyrus can be associated with a more pronounced impairment in Kana pseudo words reading [6,7]. There is considerable functional neuroimaging evidence that links pseudowords reading to the LOT area. Dietz et al. [8] found the activity within the fusiform gyrus was particularly associated with reading tasks demanding increased phonological decoding (reading pseudowords aloud). Graves et al. [9] also propose that this area serves a relatively specific and integrative function such as the mapping between ortography to fonology. fMRI data is also consistent with a functional heterogeneity of the fusiform gyrus while reading. The posterior region of the fusiform gyrus seems to be mostly involved in sublexical decoding, as evidenced by its activation with bigrams and word anagrams and its functional connectivity with prefrontal areas while reading pseudo words [10].

Our patient has a deficit with a predominant involvement of pseudo-word reading. This fact, along with all the literature reviewed, seems to indicate a distinct pathway for phonological processing within the area of fusiform gyrus. The alternative hypothesis could be a reading deficit merely dependent upon the increased complexity that pseudowords pose to a common underlying reading system. Parallel distributed processing models of reading postulate a single mechanism that generates pronunciations for all words, which are learned through repeated training within a corpus of written and spoken inputs [1]. Pseudo words would constitute letter strings that carry novelty and a complete absence of previous strengthening in connections, thus requiring more widespread areas of activation, with recruitment of similar prelexical arrays to assist their phonological decoding. A double dissociation between the phonological and lexical/semantic pathways in Western language would have to be described with basal LOT lesions to fully claim that these pathways are dissociable in this particular area. Recently, Price & Devlin [11] proposed that the LOT area is involved in reading as an interactive area, where the synthesis of visual inputs carried in the forward connections, top-down predictions conveyed by backward connections, and the mismatch between these bottom-up and top-down inputs are carried out. This model explains the higher activation of this area in pseudo words reading relative to words because of increased prediction error. The findings in our patient can also be understood according to this framework.

Finally, some relevant clinical implications can be drawn. It is not possible to know, in the patient described here, if this type of test can predict post surgical deficits, because the putative functional relevant area was not removed. Other studies in epileptic patients have demonstrated that the functional deficits (speech arrest) triggered by electrical stimulation are not always associated with post surgical deficits [12]. However, a previous report has shown that pure alexia with letter by letter reading can be surgically-induced by lesioning the basal LOT area [13]. Until these uncertainties are solved, brain mapping of his area should include reading. Specifically, tasks sensitive for different types of reading disorders may be important for functional mapping of the basal LOT cortex (Table 1). The errors listed under electrode 6 incorporate all bipolar stimulations that included this electrode. The few errors detected upon stimulation of the other electrode pairs were consistent with the patient's pre-stimulation deficits and were also verified without stimulation (Figure 1).

Table 1: Errors elicited upon bipolar stimulation of the electrodes grid.

Tasks	Errors	
Reading tasks	**Electrode 6**	**Other electrodes**
Words	5/13 (38%)	0/61 (0)
Pseudowords	3/3 (100%)	2/13 (15%)
Sentences	6/11 (54%)	1/42 (2%)
Numbers	2/3 (67%)	0/7 (0)
Other tasks		
Picture description	0/4 (0%)	3/31 (10%)
Colour naming	0/6 (0%)	1/19 (5%)
Drawings naming	0/5 (0%)	4/33 (12%)
Familiar faces naming	0/2 (0%)	6/15 (40%)
Calculus	0/3 (0%)	0/2 (0%)

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